



Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

Ultrasonographic findings of thyroglossal duct papillary carcinoma: A case report



Chonticha Srivanitchapoom^a, Pichit Sittitrai^{b,*}, Kedsaraporn Yata^a,
Piyadara Khongpiboonkit^c

^a Otolaryngology Unit, Phayao Hospital, Phayao, 56000, Thailand

^b Department of Otolaryngology, Chiang Mai University, Chiang Mai, 50200, Thailand

^c Radiology Unit, Phayao Hospital, Phayao, 56000, Thailand

ARTICLE INFO

Article history:

Received 24 December 2016

Received in revised form 2 February 2017

Accepted 5 February 2017

Available online 13 February 2017

Keywords:

Thyroglossal duct cyst

Thyroglossal duct cyst carcinoma

Sistrunk's operation

ABSTRACT

INTRODUCTION: Reports on thyroglossal duct cyst carcinoma (TGDCCa) are rare, occurring in approximately 1% of thyroglossal duct cyst (TGDC) cases. The origin and treatment of carcinoma arising in TGDC are controversial.

PRESENTATION OF CASE: A 38-year-old woman presented with a midline neck mass at the thyrohyoid level for 3 years. Ultrasound revealed a 2.4 cm cystic mass with a solid mural component and microcalcification. A small right thyroid nodule was also detected. Sistrunk's operation was performed and the pathology was a primary carcinoma arising in the TGDC with a close surgical margin. Total thyroidectomy was done and revealed a 4 mm papillary carcinoma with partial invasion through the thyroid capsule of the right lobe with a 1 mm papillary carcinoma at the isthmus. The diagnosis was a primary TGDCCa with multifocal papillary thyroid carcinoma.

DISCUSSION: Sistrunk's operation is an accepted procedure for the treatment of both TGDC and TGDCCa. Additional total thyroidectomy has been proposed but still controversial. The aims of preoperative ultrasound and ultrasound-guided fine needle aspiration biopsy (FNAB) are differential diagnosis of the possible diseases and operative planning. The results which suggest a carcinoma arising in the TGDC, synchronous thyroid malignancy and metastatic cervical lymph nodes are helpful in determining the magnitude of the operation.

CONCLUSION: Ultrasound and FNAB of the TGDC, thyroid gland and cervical lymph nodes are the useful preoperative evaluations leading to the accurate diagnosis. The definitive treatment is Sistrunk's operation with the possible addition of total thyroidectomy and neck dissection when indicated.

© 2017 The Author(s). Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

1. Introduction

The most common congenital neck mass found in over 75% of cases of childhood midline neck mass is the thyroglossal duct cyst (TGDC) [1,2] and it also has been reported in approximately 7% of the adult population [3,4]. Thyroglossal duct cyst carcinoma (TGDCCa) is very rare, accounting for only 0.7–1.6% of the cysts [5–8]. The differential diagnosis between TGDC and TGDCCa is difficult because both diseases have a similar clinical manifestation, specifically an asymptomatic midline neck mass [7,9,10]. A sudden enlargement of the mass can occur during infection however if the mass is fast growing, of a hard consistency, is fixed to surround-

ing structures and there is evidence of cervical lymphadenopathy a malignancy should be suspected [1,3,5,6].

An accurate diagnosis of TGDCCa is usually achieved from histopathology after surgical excision [5,7]. Following diagnosis, Sistrunk's operation which involves en bloc mass excision and central hyoidectomy with tract excision up to the foramen caecum is recommended for TGDCCa [9]. In addition, total thyroidectomy, neck dissection and radioiodine ablation have been described in treatment planning [2,7]. A preoperative diagnosis of TGDCCa is helpful in planning the extent of the surgery, the selection of additional therapy and the information shared with the patients [5,6,9].

2. Presentation of case

A 38-year-old woman presented with a painless midline neck mass for 3 years. She had no associated symptoms and no history of previous neck irradiation. She was clinically euthyroid. Examination revealed a 2 × 2 cm cystic mass in the thyrohyoid area.

* Corresponding author.

E-mail addresses: renoamay@hotmail.com

(C. Srivanitchapoom), psittitrai@yahoo.com (P. Sittitrai), katekoy.ent@hotmail.com (K. Yata), kpiyadara@hotmail.com (P. Khongpiboonkit).

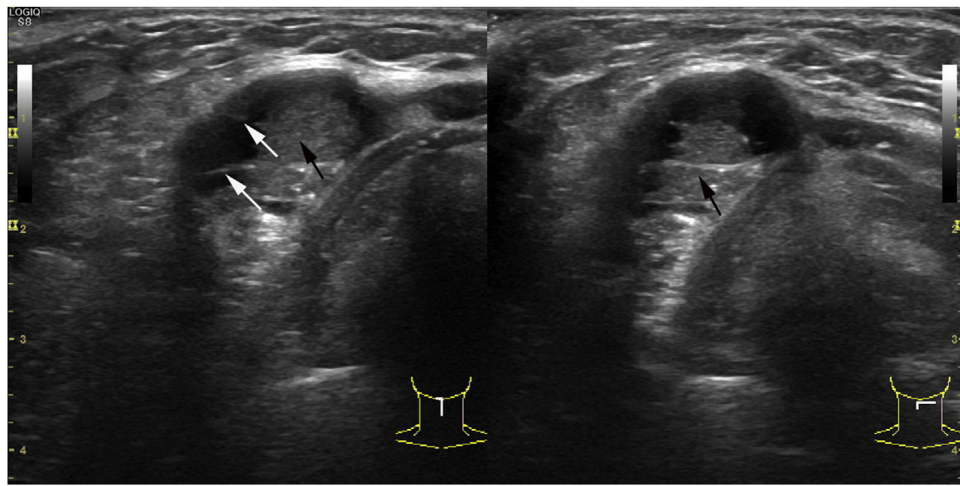


Fig. 1. A midline cystic mass with septation (white arrow) and a solid mural component (black arrow).

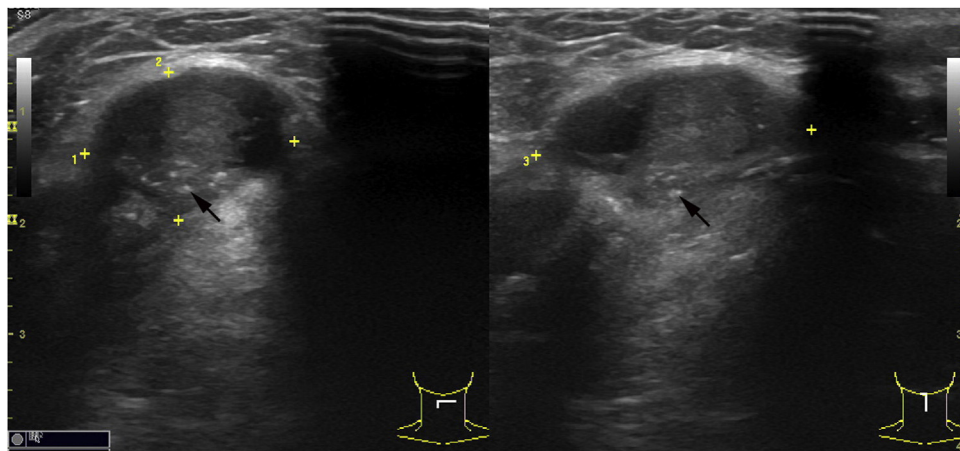


Fig. 2. Microcalcification (black arrow) in the mural mass.

The palpable thyroid gland was unremarkable. Preoperative ultrasound revealed a 1.8×2.4 cm cystic mass with septation and a solid mural component with microcalcification (Figs. 1 and 2). A 4 mm right thyroid nodule was also detected (Fig. 3) and there was no cervical lymphadenopathy. A fine needle aspiration biopsy without imaging guidance was carried out and yielded a nondiagnostic result. Sistrunk's operation was performed and the pathological report was a 1.4 cm papillary thyroid carcinoma in the benign cyst. A primary carcinoma arising in the TGDC is suggestive due to the presence of a thick fibrous wall with squamous epithelium and normal follicular cells. Lymphovascular invasion and a close surgical margin were reported. The patient was informed for a total thyroidectomy due to the pathological results and the right lobe lesion. Pathological report of the thyroid was a 4 mm papillary carcinoma with partial invasion through the thyroid capsule of the right lobe with a second 1 mm papillary carcinoma at the isthmus. The final diagnosis was a primary carcinoma arising in the TGDC with multifocal papillary thyroid carcinoma. Radioactive iodine ablation was then administered. At the 12 month follow up involving clinical examination, cervical ultrasound and radioactive iodine whole body scan there was no evidence of any recurrent disease. The thyroglobulin and antithyroglobulin antibody levels were <0.2 ng/mL and <20 IU/mL respectively. The patient has been taking synthetic thyroid hormone with a thyroid stimulating hormone level of 0.5 mU/L.

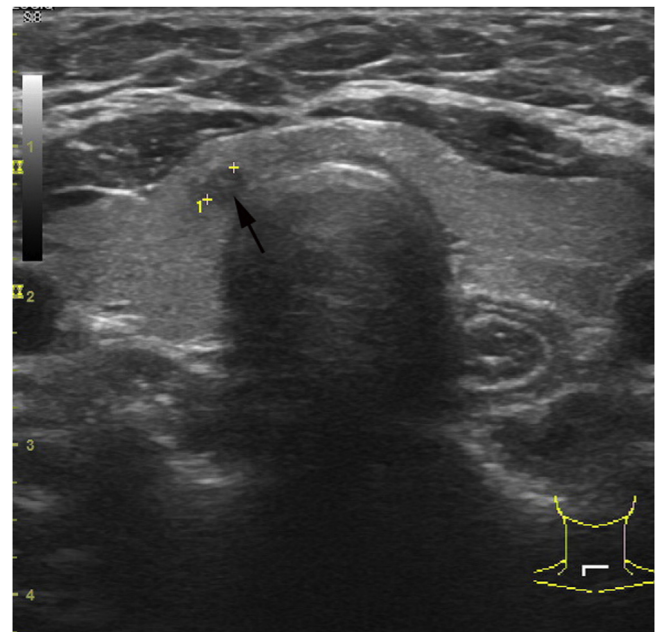


Fig. 3. A small hypoechoic right thyroid nodule (black arrow).

Table 1
Suspicious characters for thyroglossal duct cyst papillary carcinoma by imaging.

Ultrasound
Cystic with solid component [5,6,9]
Intracystic septation [14]
An echogenic mural mass with intralesional punctate calcifications (microcalcifications) [5,9,14]
Cervical lymphadenopathy [9,14]

3. Discussion

Carcinoma arising in the TGDC typically presents as a midline cystic neck mass which is identical to the benign TGDC and diseases of the pyramidal lobe of the thyroid (including colloid nodules, follicular adenoma, cystic Hashimoto's thyroiditis, thyroid carcinoma, and ectopic thyroid tissue), cystic parathyroid tumors, cystic papillary thyroid carcinoma metastasizing to Delphian lymph nodes, dermoid cysts, and branchial cleft cysts located in the midline [4,8,10]. Due to its rarity, fewer than 200 cases have been reported in literature published in English [2,3,5,7,10]. The diagnosis of TGDCa is usually made from surgical specimens of an excised TGDC [8].

Preoperative imaging is aimed at 1) performing the differential diagnosis of neck masses 2) checking characteristics of any malignancy in the mass 3) evaluating the anatomically functional thyroid gland and 4) guidance for the specific site of the tissue biopsy [2,5,8]. Ultrasonography (US) is the imaging test of choice due to the advantage of being simple, rapid, inexpensive, and does not involve any exposure to radiation. US highlights suspicious features of a carcinoma and can be used for imaging guidance for a fine needle aspiration biopsy (FNAB) [9,11–14]. An uncomplicated TGDC from US images may either show an anechoic, well circumscribed cyst or a pseudo solid mass due to the presence of proteinaceous fluid content whilst TGDCa may appear as a cystic mass with a solid component on the wall (mural mass), sometimes with evidence of microcalcification, or as a tumor invading the cyst wall [1,5,6,8,9,14] (Table 1).

Due to the simplicity of the procedure and the low complication rate, preoperative diagnostic FNAB has been recommended for use in adult patients for treatment planning and patient counseling and also for enabling a differential diagnosis from other malignant diseases such as lymphoma and metastatic Delphian lymph nodes [3,4,8,11,12]. However, because of the low frequency of malignancy in children, routine FNAB may not be cost-effective and appropriate [3]. The diagnostic accuracy has been reported as only 53% with a false negative rate of 47% due to the hypocellularity of the cystic content [3,8]. The patient in this study did not have an ultrasound-guided FNAB of the midline mass and the thyroid mass so a second operation was needed. From this we recommend cytological sampling of the solid component and the suspicious thyroid nodule using ultrasound-guided FNAB and on-site cytologic evaluation which may improve future outcomes.

Carcinoma in TGDC can be classified as primary, secondary and multicentric [5]. Primary TGDCa is that the carcinoma arises de novo from the cyst [3]. The evidence to support this theory is the cell lining of the TGDC is typically squamous epithelium but heterotrophic thyroid tissue has been identified in situ up to a level of 62% [2,5,8]. Secondary TGDCa is that the carcinoma metastasizes from an occult primary tumor in the thyroid gland because carcinoma has been observed in 20–56% of the removed thyroid gland in TGDCa cases [3,9,11–13]. In multicentric TGDCa, the histologic report of the resected midline mass should be a follicular cell-derived carcinoma with the adjacent area of normal follicular cells and epithelial cells whilst the thyroid gland also has the second primary carcinoma [5].

The definitive management of TGDCa is Sistrunk's operation [2,8]. Opinion on whether addition of a total thyroidectomy and prophylactic neck dissection to Sistrunk's operation should be carried out is still controversial [2,3,13]. Although It has been shown that these additional procedures have no significant effect on overall survival [2,13], the incidence of papillary thyroid carcinoma (PTC) in TGDCa cases when the thyroid gland has been removed is up to 56% [2,3,8,12] and cervical lymph node metastasis from PTC of the TGDCa has been reported in 16–25% of cases [3,13]. We recommend these surgical extensions should not routinely apply to all TGDCa patients. US should be carried out in every patient, not only to evaluate the midline cystic mass but also to enable the investigation of the thyroid and cervical lymph nodes. The sonographic characteristics which are suggestive of malignancy in the detected thyroid nodules are solid hypoechoic nodules with microcalcification, irregular/ill-defined margins, rim calcification with a small extrusive soft tissue component, extrathyroid extension and taller than wide [15]. US features of the lymph nodes suspected of metastatic thyroid malignancy are microcalcification, cystic aspect, peripheral vascularity, hyperechogenicity and round shape [15]. FNAB of the suspected thyroid nodule and lymph node should be carried out to ensure accurate diagnostic confirmation. The criteria for a thyroidectomy include malignancy or suspicion of malignancy from the cytologic report of a thyroid nodule size >1 cm, extrathyroidal extension of the carcinoma, nodal metastasis, a history of head and neck irradiation, and a family history of thyroid cancer [15]. Thyroidectomy is also mandated if TGDCa has a close or positive pathological margin or there is evidence of a multifocal disease, for example TGDCa with synchronous thyroid carcinoma as the patient in this study. Therapeutic central and lateral compartment neck dissection is recommended for patients with clinically metastatic nodes. Radioactive iodine (RAI) may also be considered in cases of patients of intermediate to high risk of recurrence such as extrathyroidal extension, extracapsular extension of TGDCa, lymph node metastasis, or distant metastasis [10,15]. Thyroid hormone therapy is prescribed generally at a dose necessary to keep the thyroid stimulating hormone (TSH) level being suppressed [12,15]. The overall prognosis of TGDCa is excellent however long term follow up is recommended [4,7,11,13]. Periodic clinical examination and US for surveillance are significantly useful and serum thyroglobulin needs to be evaluated during follow up in patients who have undergone a total thyroidectomy with or without RAI [10,12,15].

4. Conclusion

TGDCa should be considered a possibility in TGDC patients presenting with a hard mass which is fixed to the surrounding tissue or associated with cervical lymphadenopathy. Ultrasound of the mass, thyroid gland and cervical lymph nodes is the most useful preoperative evaluation in every age group. The definitive treatment of TGDC and TGDCa is Sistrunk's operation but if the ultrasound reveals the signs of carcinoma in TGDC and thyroid mass, these could be either secondary or multifocal TGDCa, then the addition of total thyroidectomy should be considered.

Conflicts of interest

None of the authors have any conflicts of interest to disclose.

Funding

None.

Ethical Approval

This research study was reviewed and approved by Ethic committee of Phayao hospital, Phayao, Thailand with the study approval code of HE-02-60-0008.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Dr. Chonticha Srivanitchapoom and Dr. Pichit Sittitrai were involved in writing the paper, data collection and preparing the literature review. Dr. Kedsaraporn Yata was involved in data collection and preparing the literature review. Dr. Piyadara Khongpiboonkit was involved in radiographic data analyses. All authors approved the final version of the manuscript.

Guarantor

Chonticha Srivanitchapoom, M.D. and Pichit Sittitrai, M.D. response for this work.

References

- [1] R.A. Dedivitis, D.L. Camargo, G.L. Peixoto, L. Weissman, A.V. Guimarães, Thyroglossal duct: a review of 55 cases, *J. Am. Coll. Surg.* 194 (2002) 274–277.
- [2] S.G. Patel, M. Escrig, A.R. Shaha, B. Singh, J.P. Shah, Management of well-differentiated thyroid carcinoma presenting within a thyroglossal duct cyst, *J. Surg. Oncol.* 79 (2002) 134–139, discussion 140–1.
- [3] Y.M. Choi, T.Y. Kim, D.E. Song, S.J. Hong, E.K. Jang, M.J. Jeon, et al., Papillary thyroid carcinoma arising from a thyroglossal duct cyst: a single institution experience, *Endocr. J.* 60 (2013) 665–670.
- [4] Y.J. Yang, S. Haghir, J.R. Wanamaker, C.N. Powers, Diagnosis of papillary carcinoma in a thyroglossal duct cyst by fine-needle aspiration biopsy, *Arch. Pathol. Lab. Med.* 124 (2000) 139–142.
- [5] M. Cignarelli, A. Ambrosi, A. Marino, O. Lamacchia, R. Cincione, V. Neri, Three cases of papillary carcinoma and three of adenoma in thyroglossal duct cysts: clinical-diagnostic comparison with benign thyroglossal duct cysts, *J. Endocrinol. Invest.* 25 (2002) 947–954.
- [6] N.R. Aculate, H.B. Jones, A. Bansal, M.W. Ho, Papillary carcinoma within a thyroglossal duct cyst: significance of a central solid component on ultrasound imaging, *Br. J. Oral Maxillofac. Surg.* 52 (2014) 277–278.
- [7] R. Dzodic, I. Markovic, B. Stanojevic, V. Saenko, M. Buta, I. Djuricic, et al., Surgical management of primary thyroid carcinoma arising in thyroglossal duct cyst: an experience of a single institution in Serbia, *Endocr. J.* 59 (2012) 517–522.
- [8] C.P. Plaza, M.E. López, C.E. Carrasco, L.M. Meseguer, L. Perucho Ade, Management of well-differentiated thyroglossal remnant thyroid carcinoma: time to close the debate? Report of five new cases and proposal of a definitive algorithm for treatment, *Ann. Surg. Oncol.* 13 (2006) 745–752.
- [9] A.T. Ahuja, K.T. Wong, A.D. King, E.H. Yuen, Imaging for thyroglossal duct cyst: the bare essentials, *Clin. Radiol.* 60 (2005) 141–148.
- [10] Y. Carter, N. Yeutter, H. Mazeh, Thyroglossal duct remnant carcinoma: beyond the Sistrunk procedure, *Surg. Oncol.* 23 (2014) 161–166.
- [11] M. Tharmabala, R. Kanthan, Incidental thyroid papillary carcinoma in a thyroglossal duct cyst – management dilemmas, *Int. J. Surg. Case Rep.* 4 (2013) 58–61.
- [12] D.M. Hartl, A. Al Ghuzlan, L. Chami, S. Leboulleux, M. Schlumberger, J.P. Travagli, High rate of multifocality and occult lymph node metastases in papillary thyroid carcinoma arising in thyroglossal duct cysts, *Ann. Surg. Oncol.* 16 (2009) 2595–2601.
- [13] W. Kermani, M. Belcadhi, M. Abdelkéfi, K. Bouzouita, Papillary carcinoma arising in a thyroglossal duct cyst: case report and discussion of management modalities, *Eur. Arch. Otorhinolaryngol.* 265 (2008) 233–236.
- [14] L. Solbiati, V. Osti, L. Cova, M. Tonolini, Ultrasound of thyroid, parathyroid glands and neck lymph nodes, *Eur. Radiol.* 11 (2001) 2411–2424.
- [15] B.R. Haugen, E.K. Alexander, K.C. Bible, G.M. Doherty, S.J. Mandel, Y.E. Nikiforov, et al., American thyroid association management guidelines for adult patients with thyroid nodules and differentiated thyroid cancer, *Thyroid* 2016 (26) (2015) 1–133.

Open Access

This article is published Open Access at sciendoirect.com. It is distributed under the [IJSCR Supplemental terms and conditions](#), which permits unrestricted non commercial use, distribution, and reproduction in any medium, provided the original authors and source are credited.